VASCULAR AND ENDOVASCULAR TECHNIQUES

Peter F. Lawrence, MD, Section Editor

Operative technique for tracheo-innominate artery fistula repair

Kojiro Furukawa, MD, Keiji Kamohara, MD, Manabu Itoh, MD, Hiroyuki Morokuma, MD, and **Shigeki Morita, MD,** *Saga, Japan*

Tracheo-innominate artery fistula is fatal unless treated surgically. We describe our surgical approach and results in seven patients. The average patient age was 15.7 years; all patients had prior severe neurological deficits. Three of seven patients were in hemorrhagic shock; control of preoperative bleeding was achieved with tracheostomy tube cuff overinflation. The innominate artery and the trachea were exposed through a collar incision and partial upper sternotomy. The innominate artery was divided at the aortic arch and at the bifurcation, with one exception. Cerebral blood flow was monitored by the blood pressure difference in the bilateral upper extremities and by near-infrared spectroscopy. The tracheal fistula was left adherent to the innominate artery in all but one patient. All patients were discharged without new neurologic deficits or severe morbidity. Overall survival was 84% at 37 months, without any vascular, tracheal, or neurological events. (J Vasc Surg 2014;59:1163-7.)

A rare, but serious complication of tracheostomy is the development of a trachea-innominate artery fistula (TIF). This condition is inevitably fatal unless treated surgically, and even after treatment, the morbidity and mortality remain high. Early diagnosis is difficult, as is appropriate perioperative management and the surgical procedure itself.[1] Although there are some case reports of successful treatment, there are few clinical research papers dealing with the condition,[2,3] and no studies have reported long-term outcomes after surgery. We investigated whether our surgical technique is an appropriate treatment for TIF, based on patients' short- and long-term results.

METHODS

Patient characteristics. Emergency surgery was performed in seven patients with TIF between May 2003 and September 2011. The mean patient age was 15.7 years (range, 5-32 years); three patients were male, and four were female. All patients had severe neurological deficits prior to the appearance of TIF: three patients had cerebral palsy, one had myoclonus epilepsy, one had agenesis of the corpus callosum, one had cerebral contusion, and one had muscular dystrophy. The mean duration between tracheostomy and the diagnosis of TIF was 48.6 months (range, 7-143 months). Herald bleeding was recognized in all patients; three patients experienced hemorrhagic shock (Table I).

TIF diagnosis. The diagnosis of TIF was made based on clinical and computed tomography (CT) findings. Patients had sudden arterial bleeding from a tracheostomy tube and the radiographic finding of innominate artery compression by the trachea.

Operative technique. Preoperative hemostasis was achieved in the emergency department in all patients with over-inflation of a tracheal tube cuff (GB Adjustfit Tracheostomy Tube; Fuji Systems Corporation, Tokyo, Japan); the tracheal cannula consisted of a wired silastic tube with an adjustable wing (Fig 1). The inflation of the tracheostomy balloon was done blindly. There was no set volume instilled in the balloons; they were inflated until the bleeding stopped (Fig 1). After achieving temporary hemostasis and reaching a definitive diagnosis, emergency surgery was performed. A right supraclavicular collar incision and an upper median sternotomy was performed, with extension into the right second or third intercostal space. The innominate artery, the right common carotid artery, the right subclavian artery, and the trachea were exposed (Fig 2), and the location of the TIF was confirmed. Cerebral blood flow was monitored using the blood pressure difference between the bilateral radial arteries and by the use of regional cerebral oxygen saturation (rSO_2), as determined by near-infrared spectroscopy (TOS-96 Brain Oximeter; TOSTEC Co Ltd, Tokyo, Japan). A 3-minute test clamp of the innominate artery was used to perform these tests. The innominate artery was divided, both proximally and distally relative to the fistula, if there was no evidence of significant laterality (ie, the systolic pressure difference was within 30 mm Hg, and the rSO_2 within

From the Department of Thoracic and Cardiovascular Surgery, Faculty of Medicine, Saga University.

Author conflict of interest: none.

Reprint requests: Kojiro Furukawa, MD, Department of Thoracic and Cardiovascular Surgery, Faculty of Medicine, Saga University, 5-1-1 Nabeshima, Saga 849-8501, Japan (e-mail: furukako@cc.saga-u.ac.jp).

The editors and reviewers of this article have no relevant financial relationships to disclose per the JVS policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

0741-5214/$36.00

http://dx.doi.org/10.1016/j.jvs.2013.09.013

Table I. Preoperative patient characteristics

Patient	*Age, years/gender*	*Predisorder*	*Tracheostomy, months*	*Prebleeding*	*Shock*
1	10/female	Myoclonus epilepsy	40	Yes	No
2	16/male	Cerebral palsy	60	Yes	Yes
3	5/female	Agenesis of the corpus callosum	7	Yes	No
4	5/female	Cerebral palsy	5	Yes	Yes
5	13/female	Cerebral contusion	12	Yes	Yes
6	29/female	Cerebral palsy	73	Yes	No
7	21/male	Muscular dystrophy	143	Yes	No
Mean	15.7		48.6		

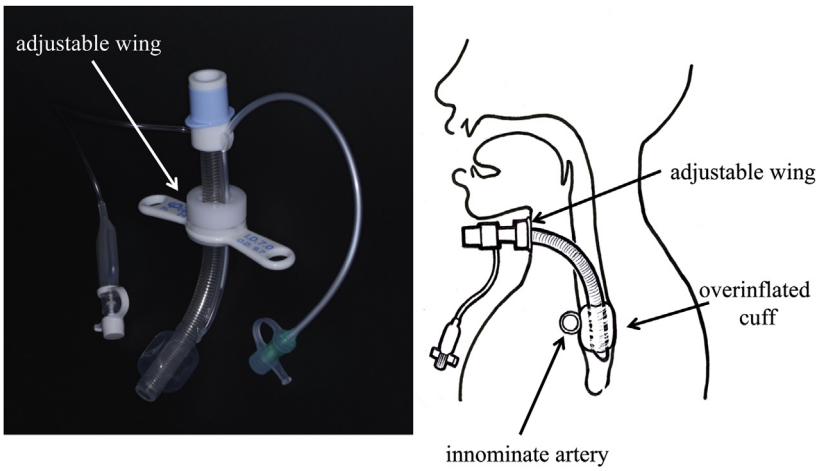

Fig 1. Tracheal tube for hemostasis. A tracheal tube used for hemostasis (*left*) and a schematic of this method obtaining temporary hemostasis (*right*).

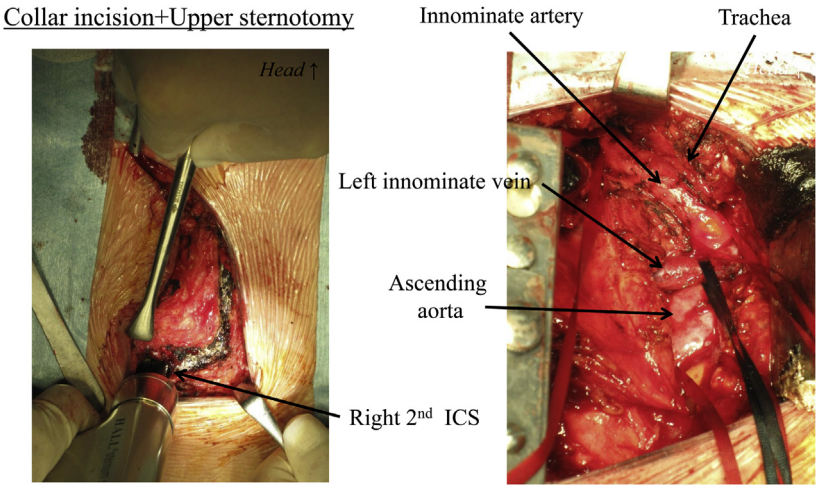

Fig 2. Operative findings. A right supraclavicular collar incision and upper median sternotomy with extension into the right second intercostal space (*ICS, left*). The innominate artery, ascending aorta, and trachea are exposed (*right*).

10%). Both ends of the divided artery were oversewn with a running 5-0 polypropylene suture, carefully maintaining the continuity between the right common carotid and the subclavian artery. If the systolic radial pressure and rSO_2 showed laterality, we would have performed innominate-to-innominate or innominate-to-right common carotid artery bypass. As a general principle, the intervening arterial segment was not dissected off the anterior wall of the trachea but rather used for tracheal closure plasty. Adjacent viable tissue (thymus or adipose tissue) was used to cover all sutures

Table II. Operative results

Patient	*Innominate artery*	*Tracheal fistula*	*Neurological complication*	*Head CT*	*Cerebral blood scintigraphy*
1	Division	Patch closure	None	No change	NA
2	Division	Left with artery	None	No change	NA
3	Division	Left with artery	None	No change	*Rt* ↓
4	Division	Direct closure	None	No change	*Rt* →
5	Division	Left with artery	None	No change	NA
6	Division	Left with artery	None	NA	NA
7	Bypass to right common carotid artery	Left with artery	None	NA	NA

CT, Computed tomography; *NA,* not applicable.

and tracheal defects. In one patient with evidence of decreased cerebral blood flow on clamping, a bypass from the innominate artery to the right carotid artery, rather than arterial division, was performed; an expanding polytetrafluoroethylene graft was used. Mechanical ventilation was continued through the tracheostomy tube, with the cuff positioned below the fistula. Patients were transferred to the intensive care unit after surgery, where they remained until their respiratory condition was stable.

Follow-up. The follow-up rate was 100%, with a mean follow-up period of 37 months (range, 3-103 months). All patients were followed with routine clinical examinations after hospital discharge.

Statistical analysis. Long-term survival was calculated using the Kaplan-Meier method, using Statistical Package for the Social Sciences (SPSS) software, version 19 (SPSS; IBM, Armonk, NY).

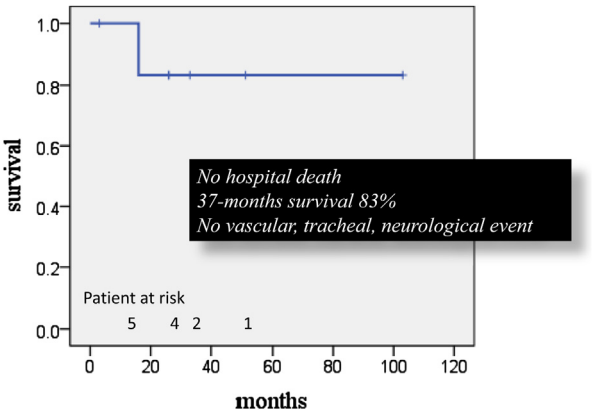

Fig 3. Long-term results.

RESULTS

Operative results. The innominate artery was divided in six patients. A bypass from the innominate artery to the right carotid artery was performed in one patient who had a 30 mm Hg decrease in the right upper extremity after the innominate artery test clamp; this patient had muscle dystrophy with no defects in consciousness. In six patients, the tracheal fistula was left adherent to the innominate artery; the tracheal defect was closed with a pericardial patch in the remaining patient. All patients were discharged without new neurological complications. In four patients, head CT verified that there were no new abnormal findings. Cerebral blood flow scintigraphy was performed in two patients; one demonstrated no laterality in cerebral blood flow, and the other had slightly decreased flow to the right hemisphere (Table II).

Long-term results. One patient died of acute pancreatitis 16 months after TIF repair. The survival rate was 83% at 37 months (the mean follow-up period). No patients experienced vascular, tracheal, or neurological events during the follow-up period (Fig 3).

DISCUSSION

Although there have been many case reports of early-onset TIF dating from the 1970s, some more recent reports have described patients with late-onset TIF; in particular, TIF occurring after laryngotracheal separation and tracheostomy in patients with severe neuromuscular disorders.[4] In patients with neuromuscular disorders, the conditions of tracheostomy, long-term mechanical ventilation, and spinal deformities are known to contribute to erosion in the tracheal wall and subsequent TIF.[5] The incidence of late-onset TIF is likely to increase as treatment for patients with severe neuromuscular disorders improves, and the patients' life spans subsequently increase. It is therefore important to establish a definitive treatment strategy for TIF.

TIF outcomes are largely dependent upon a timely diagnosis, immediate control of hemorrhage, maintenance of a patent airway, and the type of repair performed. The diagnosis of TIF is based on clinical suspicion; arterial bleeding in patients with tracheostomies is a key diagnostic feature. Nonlife-threatening premonitory airway bleeding may precede massive bleeding and should not be missed, as any delay in diagnosis puts the patient at risk for recurrent bleeding and exsanguination. CT findings, in which the innominate artery is displaced by the trachea, confirm the diagnosis of TIF.

Controlling hemorrhage and maintaining a patent airway are the goals of initial treatment. In a report of 37 patients, 12 died because of hemorrhage control and airway security failure, giving a preoperative mortality of 32%.[1] Various techniques have been reported to control

bleeding in patients with TIF. We used a tracheostomy cannula with a wired silastic tube and an adjustable wing, adjusting the position of the cuff and using cuff over-inflation to provide hemostasis. We were able to control the curvature of the tube and the length between the wing and the tip of the cannula[6]; temporary hemostasis was achieved in all patients.

Various surgical approaches are employed in TIF repair. The most commonly used incision is a median sternotomy, although two other approaches have been reported: an upper median sternotomy with extension into the right third or fourth intercostal space, extending the tracheostomy wound to the right,[2,7] and a right anterior third-interspace thoracotomy with a separate neck incision.[7] Although a full median sternotomy provides the best exposure for the entire supra-aortic trunk, the approach also carries the risk of both mediastinitis and graft infection; a 39% incidence of sternal wound complications has been reported in patients with tracheostomy and median sternotomy.[8] Our method, a right supraclavicular collar incision and upper median sternotomy, with extension into the right second or third intercostal space, is sufficient for exposure of the entire innominate artery and trachea and carries a much lower risk of mediastinitis and graft infection than full median sternotomy. None of our patients experienced infectious complications.

Various methods have been reported to deal with the innominate artery, including resection or ligation of the artery[1,3,7] and reconstruction of the antegrade cerebral blood flow using an anatomical s[2,7,9] or extra-anatomical bypass. Maintenance of blood flow can be accomplished either with direct repair of the defect or by interposition grafting. There are reports of fatal outcomes due to re-bleeding after attempts to preserve the flow in the innominate artery, using direct sutures or prosthetic material in an infected area.[1,10] In addition, it is possible that these methods could introduce critical graft infection and subsequent TIF recurrence. Interruption of flow is another option, and is accomplished by simple ligation, division, or resection of the innominate artery while attempting to preserve the right carotid-right subclavian junction. Ligation or division of the innominate artery is associated with a risk of brain ischemia; we therefore routinely monitor bilateral radial artery pressure and cerebral blood flow using near-infrared spectroscopy. This is a simple, noninvasive, and reproducible method for monitoring cerebral perfusion that provides continuous data. Samra et al reported that a 20% decrease in rSO_2 from the preclamp value is the cutoff point at which the patient is deemed at risk for cerebral ischemia with carotid artery cross-clamping.[11] Carotid stump pressure monitoring has been reported as an alternative method of determing cerebral blood flow, and a significant correlation between rSO_2 and this pressure measurement has been determined.[12] We confirmed that no significant right cerebral blood flow reduction existed in all but one of our patients before proceeding with division of the innominate artery; the single patient with evidence of decreased cerebral blood flow underwent a bypass from the innominate artery to the right carotid artery, using a polytetrafluoroethylene graft reinforced with rings. In all patients, including the six with innominate artery division, no new neurologic deficits and no new abnormal CT findings were recognized either postoperatively or during long-term follow-up. These findings confirm that our method of innominate artery division is safe and appropriate for TIF repair. Gelman et al[10] reported 19 TIF survivors who underwent operative intervention maintaining flow through the innominate artery; only 15.8% survived long-term. They also described 52 TIF survivors who underwent procedures that interrupted flow through the innominate artery; 71.2% survived long-term. These data support our findings.

Controversy exists as to the appropriate management of tracheal stomata after resection or ligation of the innominate artery.[10] Some authors propose that the stoma be left unrepaired,[3,13] while others have advocated direct closure with a pleural, muscle, or adjacent soft tissue flap.[2,9] In acute TIF, adhesions between the innominate artery and the trachea are immature. However, in late-onset cases, the innominate artery may be severely adherent to the trachea, with inflammatory tissue interposed between the structures. The division is difficult in this case, and carries the risk of mediastinitis and graft infection. The innominate artery itself is the best material for fistula coverage, and therefore should not be divided from the fistula in late-onset situations.

In the current era of endovascular treatment, stenting of the innominate artery may be an option for TIF treatment.[14,15] The endovascular approach offers a valuable, less invasive alternative for patients with TIF. The technology of endovascular stents is evolving; the risk of infection is still present, with potentially catastrophic results including re-bleeding. The use of a stent can be a temporizing measure before a more definitive surgical procedure is carried out.

There are no detailed reports describing long-term results after surgery for TIF repair. In a review article, only 24% of 24 total patients survived 2 months postoperatively.[1] Our much better long-term results validate our surgical strategy.

The mortality for TIF is extremely high; therefore, the ideal management of TIF is its prevention. Several factors contribute to the formation of a TIF, including tracheostomies performed below the third or fourth tracheal rings, over-inflated cuffs, and positive-pressure ventilation.[2,5] These conditions should be avoided, especially in neck vessel anomalies and spinal deformations.[5]

Our study does have some limitations. Our patient population already had significant neurological compromise and potentially altered cerebral blood-flow requirements. Innominate ligation without reconstruction would preferably be performed in patients without neurological disease, however, innominate ligation is the safest method in the devastating situation of a TIF. If test clamping of the innominate artery causes a significant decrease in flow to the right

hemisphere, the innominate artery should be reconstructed with a prosthetic graft. In addition, our patients were a very young cohort and had the physiologic reserve to tolerate significant blood loss and emergency operation. This may well have been a factor in our ability to salvage all patients.

CONCLUSIONS

Preoperative temporary control of bleeding in patients with TIF is crucial; an adjustable tracheostomy tube is very useful for this purpose. Division of the innominate artery under cerebral blood-flow monitoring is safe and appropriate for definitive TIF treatment. When adhesions between the innominate artery and the trachea are severe, separation of the innominate artery from the trachea is not required.

The authors would like to thank Dr Suetsugu for creating the outstanding drawing used in Fig 1.

REFERENCES

1. Yang FY, Criado E, Schwartz JA, Keagy BA, Wilcox BR. Trachea-innominate artery fistula: retrospective comparison of treatment methods. South Med J 1988;81:701-6.
2. Jones JW, Reynolds M, Hewitt R, Drapanas T. Tracheo-innominate artery erosion: successful surgical management of a devastating complication. Ann Surg 1976;184:194-204.
3. Hafez A, Couraud L, Velly JF, Bruneteau A. Late cataclysmic hemorrhage from the innominate artery after tracheostomy. Thorac Cardiovasc Surg 1984;32:315-9.
4. Baydur A, Kanel G. Tracheobronchomalacia and tracheal hemorrhage in patients with Duchenne muscular dystrophy receiving long-term ventilation with uncuffed tracheostomies. Chest 2003;123:1307-11.
5. Iodice F, Brancaccio G, Lauri A, Donato RD. Preventive ligation of the innominate artery in patients with neuromuscular disorders. Eur J Cardiothorac Surg 2007;31:747-9.
6. Yokoyama M, Tayama N, Kurauchi T, Mizuno M, Niimi S, Hirose H. An adjustable tracheal cannula. J Jpn Bronchoesophageal Soc 1993;44:477-80.
7. Ramesh M, Gazzaniga AB. Management of tracheo-innominate artery fistula. J Thorac Cardiovasc Surg 1978;75:138-40.
8. Serry C, Bleck PC, Javid H, Hunter JA, Goldin MD, DeLaria GA, et al. Sternal wound complication: management and results. J Thorac Cardiovasc Surg 1980;80:861-7.
9. Eddine HJ, Ayed AK, Moosa AA, Sarraf NA. Graft repair of tracheo-innominate artery fistula following percutaneous tracheostomy. Interact Cardiovasc Thorac Surg 2008;7:654-5.
10. Gelman JJ, Aro M, Weiss SM. Tracheo-innominate artery fistula. J Am Coll Surg 1994;179:626-34.
11. Samra SK, Dy EA, Welch K, Dorje P, Zelenock GB, Stanley JC. Evaluation of a cerebral oximeter as a monitor of cerebral ischemia during carotid endarterectomy. Anesthesiology 2000;93:964-70.
12. Lee TS, Hines GL, Feuerman M. Significant correlation between cerebral oximetry and carotid stump pressure during carotid endarterectomy. Ann Vasc Surg 2008;22:58-62.
13. Silen W, Spieker D. Fatal hemorrhage from the innominate artery after tracheostomy. Ann Surg 1965;162:1005-12.
14. Deguchi J, Furuya T, Tanaka N, Nobori M, Seki Y, Nomura Y, et al. Successful management of tracheo-innominate artery fistula with endovascular stent graft repair. J Vasc Surg 2001;33:1280-2.
15. Palchik E, Bakken AM, Saad N, Saad WAE, Davies MG. Endovascular treatment of tracheo-innominate artery fistula: a case report. Vasc Endovascular Surg 2007;41:258-61.

Submitted Apr 23, 2013; accepted Sep 8, 2013.